SCIENTIFIC ARTICLE

Pain in Children Following Microsurgical Reconstruction for Obstetrical Brachial Plexus Palsy

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Purpose To determine the prevalence and characteristics of pain experienced by children who have had microsurgical reconstruction for obstetrical brachial plexus palsy (OBPP).

Methods A prospective case series study was conducted of 65 children aged 6 to 18 years with a diagnosis of OBPP and who had microsurgery at less than 12 months of age with nerve grafting or transfer. A total of 28 patients (43%) had upper OBPP and 37 (57%) had total OBPP. We evaluated pain using the Faces Pain Scale−Revised and the Adolescent Pediatric Pain Tool. Sensory symptoms in the affected limb were also collected. Mean age was 11.0 ± 3.3 years.

Results We evaluated 65 children. The point prevalence of pain (pain at the time of assessment) was 25%. The reported lifetime prevalence of pain (experienced anytime during life) was 66%. A total of 71% reported that the affected extremity felt different at least once in their lifetime. Average intensity of those with pain (n = 43) was 40 ± 19 mm on a 100-mm visual analog scale. Seventy percent of children reported that symptoms occurred every day or at least once a week. Anatomical distribution of pain was throughout the affected upper extremity irrespective of the severity of injury, with the exception of children with upper plexus injuries who did not report pain in their hand. Words typically used to describe neuropathic or musculoskeletal symptoms were chosen by the children to represent their pain.

Conclusions Children with OBPP who had microsurgical reconstruction commonly reported pain. These symptoms were typically frequent but were episodic and low in intensity. The descriptions of the type of pain include terms typical of both neuropathic and musculoskeletal origins. *(J Hand Surg Am. 2015;40(6):1177−1183. Copyright © 2015 by the American Society for Surgery of the Hand. All rights reserved.)*

Type of study/level of evidence Prognostic IV.

Key words Brachial plexus injury, children, teenagers, outcomes, pain.

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Our understanding of pain experienced by children with obstetrical brachial plexus palsy (OBPP) is limited. Perhaps the issue of pain in these patients has not received sufficient attention because of earlier findings that infants who had microsurgical reconstruction of the brachial plexus did not have neuropathic and chronic pain despite spinal root avulsions.[1] This contrasts with the typical experience of adult patients who may have debilitating neuropathic and chronic pain after with such injuries.[2] Bruxelle et al[2] reported that 91% of

TABLE 1. Demographic and Surgical Information

Demographics	Total (N = 65)		Upper Plexus (n = 28)		Total Plexus (n = 37)	
	n	%	n	%	n	%
Sex						
Male	26	40	15	54	11	30
Female	39	60	13	46	25	70
Affected Limb						
Left	20	31	9	32	11	30
Right	45	69	19	68	25	70
Surgery						
Microsurgery only	33	51	12	43	21	57
Microsurgery and secondary upper extremity reconstruction	32	49	16	57	16	43
Root avulsions*						
0	23	35	17	61	6	16
1	20	31	8	29	12	33
2	14	22	2	7	12	33
3	4	6	1	4	3	8
4	2	3	0	0	2	5
Missing data	2	3	0	0	2	5

*Statistically significant, $P < .05$.

adults with brachial plexus injuries had pain, 41% of whom categorized the pain as severe. Furthermore, spinal root avulsion was a key factor associated with pain immediately after injury and at follow-up. Bertelli et al[3] confirmed a relationship between root avulsion and pain in adults. In addition, that study found that the prevalence of pain was greater in adults with total plexus palsy than in those with upper plexus palsy. However, a comparison of pain outcomes in children with the standards used with adults fails to recognize that children may experience pain differently. It is important that both the timing of evaluation and the assessments used be able to capture the potentially delayed presentation of pain in these children and the appropriate expressive language used by children to describe pain.

Self-mutilation is a behavior associated with pain and sensory disturbance that has been investigated in children with OBPP. Al-Qattan[4] evaluated self-mutilation behaviors in children aged 2 to 8 years with OBPP. Six of the 127 children (5%) studied exhibited this behavior. The prevalence of self-mutilation was higher in children with total plexus (4 of 37) than upper plexus palsy. None of the children who exhibited these symptoms reported pain. McCann et al[5] added to these findings with comparative outcomes of children with and without microsurgical reconstruction of the brachial plexus in infancy. The incidence of self-mutilation was 29% in 24 children who had surgery and 1% (2 of 147) in those who did not have surgical treatment. The authors suggested that self-mutilation was a manifestation of a chronic pain syndrome or dysesthesias in the children identified. These reports indicated that the nature of neuropathic pain was not fully understood and could not be ruled out in children with OBPP.

The purposes of this study were to determine the prevalence of pain in children with OBPP and describe the characteristics of this pain. Secondarily, this study aimed to compare the prevalence of pain with the severity of brachial plexus injury.

MATERIALS AND METHODS

Our research ethics board approved this prospective case series. Inclusion criteria were children aged 6 to 18 years with a confirmed diagnosis of OBPP, who required microsurgery at less than age 12 months with nerve grafting or transfer. Children with a later traumatic brachial plexus injury, bilateral brachial plexus palsy, cognitive or developmental delay, or a recent (< 3 mo) injury to the affected or unaffected upper extremity at the time of recruitment were excluded.

We extracted the child's surgical information from the Hospital for Sick Children Brachial Plexus

TABLE 2. Prevalence of Pain (N = 65)

	Lifetime Prevalence of "Feels Different"				Point Prevalence of Pain				Lifetime Prevalence of Pain			
	Yes	%	No	%	Yes	%	No	%	Yes	%	No	%
Upper plexus	20	71	8	29	7	25	21	75	16	57	12	43
Total plexus	26	70	11	30	9	24	28	76	27	73	10	27
Total	46	71	19	29	16	25	49	75	43	66	22	34

TABLE 3. Frequency of Symptoms (N = 46)

	Total		Upper Plexus		Total Plexus	
	n	%	n	%	n	%
Every day	18	39	11	55	7	27
Weekly	14	30	4	20	10	39
Monthly	10	22	3	15	7	27
Yearly	4	9	2	10	2	8

database. The approach to microsurgical reconstruction of the brachial plexus included one or a combination of nerve grafting, transfer, and neurolyses. Secondary surgeries were musculoskeletal procedures of the shoulder, elbow, forearm, and wrist. The diagnosis of upper (C5 and C6 ± C7) versus total (C5, C6, C7, and C8 ± T1) palsy was confirmed by the operative report. The location and the number of root avulsions were recorded. Severity of disease was assessed by the type of brachial plexus injury (ie, upper versus total palsy) and number of spinal root avulsions.

We evaluated the point prevalence of pain using the Faces Pain Scale—Revised, a valid and reliable measure of intensity of pain in children.[6,7] The Faces Pain Scale—Revised measures intensity of pain with 6 faces representing increasing pain on a 6-point ordinal scale. The scale ranges from 0 to 10.

The children were asked whether they felt that the affected limb "felt different" from the unaffected limb during their lifetime. This was to determine the lifetime prevalence of the experience of discomfort or altered sensation in the affected limb. This question was phrased to ensure that any experience of altered sensation or discomfort was captured, even if the child may not describe this as pain. Children who reported that their limb felt different were asked to describe the feeling and rate the frequency of these symptoms on an ordinal scale: daily, weekly, once a month, and once a year. The children were then asked whether they had ever experienced pain in their affected and unaffected limb during their lifetime. Responses were recorded as "yes" or "no." The unaffected limb was evaluated as a control.

The Adolescent Pediatric Pain Tool (APPT) is a valid multidimensional pain measure developed for children and teens.[8–10] It is a paper-and-pencil test of self-reported pain for children aged 8 to 17 years. It includes a body outline diagram (BOD) to indicate the location of pain, a 100-mm visual analog scale anchored with the words "no pain," "little," "medium," "large," and "worse possible pain" (Word Graphic Rating Scale), and pain descriptors that include affective, evaluative, sensory, and temporal words. The APPT was administered to the children with assistance by the examiner. First, the children were asked to draw on the BOD where they experienced pain in their body. Second, the children completed the Word Graphic Rating Scale to rate the intensity of pain experienced during a typical episode. Third, the examiner read the list of pain descriptors to the children with a verbal prompt at the onset to inform the children to request clarification if they did not understand the meaning of a word.

We compared demographic and surgical data between children with upper and total plexus palsy using descriptive statistics (Table 1). A total of 28 children (43%) had upper plexus and 37 (57%) had total plexus palsy. Mean age at the time of assessment was 11.0 ± 3.3 years (range, 6–18 y). This did not significantly differ between children with upper (mean, 11.5 ± 3.4 y) and total (mean, 10.5 ± 3.2 y) plexus palsy. Documentation of age at the time of surgery in months and days was available for 61 of the 65 children. The 26 children with upper plexus injuries were operated at an average of 8.9 ± 2.4 months. In contrast, those who had total plexus injuries (n = 34) received surgery at an average of 4.5 ± 2.1 months. The timing of primary surgery was statistically different between the 2 groups (Mann–Whitney U test, $z = -5.3$; $P < .001$; $\alpha = .05$). The 2 groups also differed statistically when the cohort was stratified according to number of spinal root avulsions. Children with total plexus injuries had a higher number of spinal root avulsions than those with upper plexus injuries (Fisher exact test,

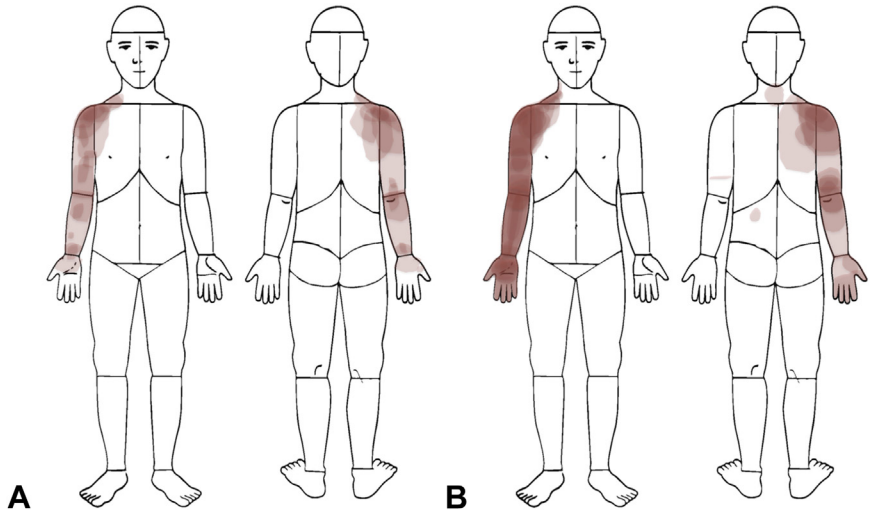

FIGURE 1: Adolescent Pediatric Pain Tool BOD. **A** Upper plexus patients. **B** Total plexus patients. For the purpose of simplifying data presentation, the drawings on the APPT BOD are represented on the right upper extremity for all patients.

$P = .001$; $\alpha = .05$). Bonferroni correction was applied when multiple comparisons were employed.

RESULTS

A total of 283 children in the Hospital for Sick Children Brachial Plexus database were eligible for the study. Seventy-three of those children attended the Hospital for Sick Children Brachial Plexus clinic during the data collection period November 2010 to September 2013. Sixty-five of these children and their families consented to this study.

Point prevalence of pain at the time of assessment was 25% (n = 16) of the 65 children evaluated (Table 2). This did not differ between those with upper plexus (n = 7, 25%) and total plexus (n = 9, 24%) palsy. Seventy-one percent of children reported that the affected arm felt different at least once in their lifetime. This experience did not differ between those with upper and total plexus palsy. The lifetime prevalence of pain was 66% (n = 43) of the total sample. The lifetime prevalence of pain in children with total plexus (n = 27, 73%) was not statistically different than in children with upper plexus (n = 16, 57%) palsy. A post hoc evaluation also demonstrated that there was no significant difference in the point and lifetime prevalence of pain in children who had microsurgery versus those who had microsurgery and one or more secondary upper extremity reconstructive procedures (Table 2). Furthermore, the 2 groups of children did not differ in their reports of whether the affected arm felt different at least once in their lifetime. Two children in this entire cohort reported pain in the unaffected limb.

At the time of assessment, the mean and SD of the Faces Pain Scale—Revised score was 0.86 ± 1.77 out of 10 in the entire sample. Average intensity of the typical pain episode in children who reported having pain at some point in their lifespan (n = 43) was 40 ± 19 mm on the 100-mm visual analog scale. This corresponds to medium pain on the World Graphic Rating Scale. The range of scores was between 1 and 81 mm. There was no statistical difference between the intensity of pain experienced at the time of assessment or during the lifetime of children with upper and total plexus palsy (Mann—Whitney U test, $z = 1.15$, $P = .270$, and $z = -0.39$, $P = .700$, respectively). Subsequently, 5 of these patients were referred to the chronic pain team for management at the authors' institution.

Seventy percent of children reported that the affected limb "felt different" every day or at least once a week (Table 3). The difference in frequency of symptoms reported between children with upper and total plexus palsy did not differ statistically (Fisher exact test, $P = .230$). Figure 1 illustrates the anatomical location of the pain experienced using the APPT BOD. For the purpose of simplifying data presentation, the drawings on the APPT BOD were represented on the right upper extremity for all patients. A comparison between children with upper and total plexus palsy is shown. Table 4 represents the anatomical location of the pain in accordance to the APPT body schemata categories. The proportion of children with pain in the hand was statistically higher in those with total plexus palsy (Fisher exact test, $P = .045$). Otherwise, the pain experienced by the whole cohort was throughout the affected upper extremity.

The APPT pain descriptors were compared in the 43 children who described pain. The percentage of

TABLE 4. Anatomical Location of Pain

	Total (N = 43)		Upper Plexus (n = 16)		Total Plexus (n = 27)	
	n	%	n	%	n	%
Anterior arm	20	47	10	63	10	37
Posterior arm	20	47	9	56	11	41
Forearm	19	44	6	38	13	48
Hand	14	33	2	13	12	44
Neck	10	23	4	25	6	22
Chest	6	14	2	13	4	15
Back	1	2	0	0	1	4
Leg	1	2	0	0	1	4

words in the evaluative, sensory, and affective categories were compared and then ranked according to the frequency of each word. Mean percentages of sensory and evaluative words reported were 21% ± 18% and 25% ± 22%, respectively. A mean of 10% ± 17% of affective words was reported. There was no statistical difference between the percentage of words chosen in the sensory and evaluative categories. However, statistically fewer affective words were chosen compared with evaluative and sensory words (Wilcoxon signed test, $z = -4.3$; $P < .001$). Table 5 ranks the APPT word descriptors with a frequency greater than the median (8.0) chosen by all children who had pain. A word cloud that represents the intensity of evaluative, sensory, and affective words is illustrated in Figure 2. The children in this study used words that are traditionally understood to represent both nociceptive or musculoskeletal pain and neuropathic pain. There was consensus that the pain experienced was episodic in nature. Words children used to describe how the limb felt different were a mixed representation with neuropathic and musculoskeletal meanings. Of the 46 children who reported that the limb felt different, 21 used words to describe symptoms suggestive of a neuropathic origin, such as "tingling," "pins and needles," "asleep," "numbness," "twitches," and "shooting pain."

TABLE 5. Adolescent Pediatric Pain Tool Word Descriptors

Evaluative, Sensory, and Affective Pain Descriptors	Frequency	Temporal Pain Descriptors	Frequency
Uncomfortable	29	Once in a while	23
Sore	22	Sometimes	20
Annoying	20	Off and on	13
Hurting	18		
Like a pinch	17		
Stiff	16		
Tight	15		
Like a hurt	14		
Uncontrollable	14		
Aching	14		
Like an ache	12		
Like a pin	12		
Cramping	11		
Hitting	10		
Like a sting	10		
Awful	10		
Numb	10		
Throbbing	10		
Shocking	9		
Like a sharp knife	9		
Pin-like	9		
Pinching	9		
Pressure	8		
Hot	8		
Sharp	8		
Crying	8		
Itching	8		
Stabbing	8		

DISCUSSION

Contrary to the early report by Anand and Birch,[1] this study demonstrated that children with OBPP requiring surgical reconstruction experience pain related to the injury in the affected upper extremity. The strength of this study was the use of both quantitative and qualitative methodologies to capture the pain experience of children with OBPP. Merely asking children about their point prevalence of pain was insufficient to capture the entire pain experience. This is illustrated by the difference between point prevalence (25%) and lifetime prevalence (62%) of pain. Evaluation of the temporal aspect of pain is important, but so is the type of pain experienced.

This study illustrates the importance of evaluating both musculoskeletal and neuropathic pain symptoms in children with OBPP. The children in this study reported pain symptoms from both etiologies. Musculoskeletal pain in children with OBPP was also reported by Spaargaren et al,[11] who prospectively interviewed children with OBPP aged 7 to 8 years about their current pain as a subcomponent of a larger

FIGURE 2: Word cloud: APPT word descriptors. The sensory, evaluative, and affective APPT word descriptors chosen by the children are illustrated. The size of the font of each word is proportionate to its frequency of report. The different categories of pain descriptors are represented in different fonts (ie, **Sensory** is in the noteworthy bold font, Evaluative is in the arial font, and Affective is in the chalkduster font).

study on activity and participation. Most of these patients studied had upper plexus injuries (n = 50; 94%). Twenty-four of the 53 patients (45%) reported pain. The location of this pain was in the shoulder (58%), upper arm and elbow (46%), and neck (38%). In this current study, the list of APPT affective and evaluative words in Table 5 appears to support these findings. Words associated with musculoskeletal pain were most frequently chosen by children with OBPP to describe pain. However, this list must be evaluated in conjunction with the qualitative findings from asking children to describe the feeling in their arm. Several of the children's own descriptive words were representative of neuropathic pain symptoms. Their own words associated with neuropathic symptoms were more often reported when asked about the sensation in the limb as opposed to pain. Consequently, not all of these children chose words on the APPT pain questionnaire that were reflective of neuropathic pain. This highlights the importance of how we ask these children about pain and how they interpret these symptoms. Children with OBPP may not necessarily describe their neuropathic symptoms as pain. This suggests that the experience of neuropathic symptoms may be different in these children because their injury occurred during infancy: at birth and from further trauma at the time of microsurgery. Many children may not label these symptoms as pain because these feelings have been integrated as a part of their everyday experience. Further research is needed to understand how children with OBPP describe neuropathic pain.

It is also important to consider how children interpret and use words to represent different types of pain. The developers of the APPT stated that sensory words such as "hot," "burning," "numbness," and "tingling" are associated with neuropathic pain.[10] Although the list of pain descriptors on the APPT was generated and validated on 958 schoolchildren, the face validity of these words to represent nociceptive or musculoskeletal pain versus neuropathic pain in children has not been established.[9,10] The words used to describe pain are insufficient to make a diagnosis regarding the etiology of the pain. A thorough history, physical examination, and possible diagnostic investigations are necessary to complete a pain diagnosis. For example, a diagnosis of impingement or rotator cuff disorder could be made with a subacromial corticosteroid injection to relieve pain from inflammation. The words chosen by the children in this study may be suggestive of one pain type or another, but they are not diagnostic. Therefore, further research is required to clearly determine the specific etiologies of pain in this population. Irrespective of the etiology of pain, the new information from this study provides evidence of the need to establish a routine clinical protocol for evaluating neuropathic and musculoskeletal pain symptoms in children with OBPP. As these needs are identified, an interdisciplinary team approach will be integral to providing optimal management. Because this study has only begun to make known the issue of pain in these children, the detailed recommendation for optimal management has yet to be determined.

Although this study was conducted with prospectively collected data, this sample was not randomly selected from the population of children who attended the clinic, because that was not feasible. Therefore, the study results must be considered within the limitations of the selection bias that must result from obtaining the sample from the authors' institution's brachial plexus

clinic. Children who routinely attend the clinic would more likely have functional impairment.

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